

Xylophagia: A meta-synthesis of the literature

ABSTRACT

Purpose: This review provides an insight into xylophagia, its treatment, intervention options, etiological causes, and possible relationship with other diseases.

Design/methodology/approach: A systematic search was performed across four scientific databases (i.e., Ovid Medline, Embase via Ovid, PubMed and ProQuest). All of the qualitative studies reporting on xylophagia from the inception of databases until August 2019 have been included. The quality of included studies was assessed through a 10-item checklist given by Kmet, Lee, and Cook in 2004.

Findings: A total of 18 studies were included, and five primary themes emerged after analysis: 1) Precipitation/onset of xylophagia, 2) Co-morbid psychiatric or medical illnesses, 3) Assessment and investigation modes to confirm diagnosis, 4) Outcomes of xylophagia, and 5) Treatment options comprising medical care, psychological care, counselling, and duration of recovery. There were 16 females and 9 males in included studies. The mean age and standard deviation of males and females were 29.25(12.17) years and 32.81(11.92), respectively. The mean duration and standard deviation of paper pica were 4.80(4.27) years.

Research implications: Despite the limitation that this meta-synthesis is based upon findings from case studies, results show that standardized medication regimens for treating xylophagia are still not available or unknown. There is a dire need for further research in order to better understand the disorder. The healthcare professionals need to use the reciprocal, mutually constituent influence of biological and sociocultural factors in order to screen, diagnose and manage problems like xylophagia.

Originality/value: The findings advance our understanding of the positive effects of patients and family members undergoing counselling or cognitive behavior therapy in reducing stress and enhancing coping skills thus avoiding self-damaging behaviors has been established.

Keywords: Xylophagia, Pica, Eating Disorder, Psychosis, Health Services, Qualitative Research, Health Policy, Quality of Care

26 **Key Points**

- 27 1. This review provides an insight into xylophagia, its treatment and intervention options, possible etiological causes,
28 postulated relationships with other diseases and highlights its inclusion in differentials of a medico-legal
29 physician.
- 30 2. This paper highlights factors that lead to precipitation/onset of xylophagia, the co-morbid psychiatric or medical
31 illnesses, various assessment and investigation modes to confirm the diagnosis of xylophagia, the outcomes of
32 xylophagia, and various treatment options comprising medical care, psychological care, counselling, and duration
33 of recovery.
- 34 3. This review includes xylophagia studies from various countries and shows that xylophagia is multifaceted and
35 underdiagnosed.
- 36 4. There were females and males in included studies indicating its prevalence across gender.
- 37 5. This review suggests a need for biopsychosocial integration in diagnosing and treating eating disorders.

38 **Introduction**

39 The simple act of eating has a certain cost as food has been shown to be the cause of millions of deaths
40 globally due to various mechanisms. The consumptions of hundreds of microbial, chemical and physical agents can
41 result in illness and have various lethal outcomes (Acheson, 1999, Jacob et al., 1990, Parakh et al., 2016, Dalal et al.,
42 2013, Barros et al., 1991). The forensic pathologists frequently come across cases of death by food and occasional
43 and culturally-specific cases of death due to intentional or unintentional intake of pathogens, poisons, and foodstuffs
44 (Byard, 2017).

45 Xylophagia is described as the consumption of substances made up of wood such as paper, pencils, boxes,
46 toothpicks, matches and tree barks; with paper ingestion the most frequently reported type (Gowda et al., 2014,
47 Moore and Sears, 1994). It is a form of pica, where a person has a developmentally inappropriate compulsive
48 yearning for and eats non-nutritive substances (Blinder and Salama, 2008). Pica is explained in the field of medicine
49 as an unnatural appetite that initiates a craving for unhealthy substances. Other kinds of pica comprise lithophagia
50 (stone eating), geophagia (dirt eating), ice (pagophagia), gravel, paint flakes, clay, hair (trichophagia), and laundry
51 starch (amylophagia) (Byard, 2014, Parry-Jones and Parry-Jones, 1992, Loots and du Toit-Prinsloo, 2016). There
52 has been much ambiguity in classification of this disorder, with the Diagnostic and Statistical Manual of Mental
53 Disorders version 5 (DSM-V) classifying it as an eating disorder in persons who fulfill the following criteria:
54 persistent eating of non-nutritive substances for a period of at least one month, the consumption of non-nutritive
55 substance that is inappropriate to the developmental level of an individual, eating behavior that is not culturally
56 supported or socially practiced, or the behavior is severe enough to call for independent clinical attention if it occurs
57 along with another mental disorder or medical condition (APA, 2013). Others, however, have suggested that it is on
58 the obsessive-compulsive disorder (OCD) spectrum, given both the obsessive and compulsive features observed in
59 patients (Bhatia and Gupta, 2009, Gundogar et al., 2003, Hergüner et al., 2008).

60 A variety of factors are involved in the etiology of pica, such as genetic, biological, and particularly
61 sociocultural and nutria-physiological aspects (Rose et al., 2000, Levine and Smolak, 2014). Likewise, a past study
62 has suggested the integration of sociocultural and biopsychiatric factors in etiology and management of eating
63 disorders (Levine and Smolak, 2014). A study conducted on pregnant women in Mexico found that between
64 approximately 30-50% of the women consumed dirt, ashes, and clay; believing that failure to do so would lead to

65 miscarriage (Simpson et al., 2000). A large part of the literature also attributes it to iron deficiency, although there
66 remains some controversy as to whether it is a cause instead of an outcome of iron deficiency (Kettaneh et al., 2005,
67 Gupta A et al., 2007). Other research studies have highlighted the role of social and religious customs, where pica is
68 considered culturally-acceptable and is thus not viewed as pathological (Blinder and Salama, 2008). This ambiguity
69 has then resulted in confusion as to how this condition should be treated. A review paper highlighting the forensic
70 implications of pica (Byard, 2014) shows that various fatal mechanisms result in the death of a person with pica. In
71 case of suspicion of pica in persons who are institutionalized or have developmental delay, post-mortem
72 examinations should cautiously analyse the possible side effects of eating non-food substances (Byard, 2014).
73 Authors highlight the presence of minimal presenting symptoms and signs due to a variety of reasons.

74 In children aged between 18 to 36 months, an incidence of pica greater than 50% is considered normal, and
75 this is thought to decrease with age (Blinder and Salama, 2008). Among institutionalized patients, prevalence rates
76 range from 4-26% (Walker and M.C. Roberts, 2001), and it was noted that from 1999 to 2009, hospitalization rates
77 due to pica increased by more than 90% (Zhao and Encinosa, 2011). The intake of foreign bodies is commonly
78 observed in children and certain susceptible adult populations such as alcoholics, the elderly with dentures,
79 prisoners, and individuals with psychiatric problems (Yamaguchi et al., 2014, Zouros et al., 2014, Cangir et al.,
80 2002, Velitchkov et al., 1996).

81 In most of the cases, the autopsy results reveal accidental intake of foreign bodies (e.g., meat or bones). In
82 rare cases, the autopsy results reveal non-food components (e.g., teeth, glass fragments, dental crowns, batteries, or
83 small toys) (Sreetharan et al., 2004), and suicidal intent must be considered when children or inebriated patients are
84 involved in ingesting these substances (Krugman et al., 2007, Žganjer et al., 2011, Bloch et al., 2005, Nicolai and
85 Reiter, 2004). There is a need for thorough knowledge and sufficient medico-legal experience in order to
86 differentiate between suicidal acts and homicide (Sauvageau and Racette, 2006). Furthermore, in rare cases of drug
87 abuse and eating disorders, there is an intake of non-food substances (Schaper et al., 2003).

88 The past empirical studies in various cases have shown that the intake of non-food pica (depending on the
89 type, number, and size of the objects) leads to adverse health outcomes (e.g., aggressive periodontitis, lead
90 poisoning, phosphorus poisoning, dental damage, mercury poisoning, nicotine poisoning, toxocara infections,
91 hemolytic anemia, electrolyte disturbances, blockage, hemorrhage, and tamponade from vessel and heart

92 perforation, and sepsis) due to failure to digest the foreign substances (McLoughlin, 1988, Decker, 1993, Johnson,
93 1990, Federman et al., 1997, Glickman et al., 1981, Toker et al., 2009, Advani et al., 2014, Kar et al., 2015, Byard,
94 2000, Byard, 2014). The past empirical evidence has shown death due to: vagal stimulation with resultant cardiac
95 arrhythmia, intestinal causes, and intestinal obstruction in some cases of pica (Wick et al., 2006, Byard, 2014). The
96 intake of hair or paper may form bezoars which lead to intestinal obstruction due to absorption of fluid in the
97 accumulated paper in the colon area (Byard, 2014). The presence of non-food substances in the aerodigestive tract of
98 children at post-mortem examination is not always indicative of pica as the children may have intentionally or
99 unintentionally eaten foreign material (Byard et al., 1990, Yamaguchi et al., 2014, Zouros et al., 2014). The variety
100 of outcomes of pica and lack of specificity has added to the problems in diagnosing pica. The cultural
101 contextualisation of eating disorders, as proposed in a past study (Levine and Smolak, 2014) needs to be
102 comprehensively assessed.

103 As it pertains to xylophagia specifically, there is a paucity of data available due to differences in definitions
104 as well as reluctance on the part of patients to admit to it (Blinder and Salama, 2008), further thwarting efforts to
105 diagnose, prevent, and manage this disease comprehensively and effectively. Hence, there is ambiguity on the
106 etiology of xylophagia and limited empirical evidence on the fatal or potentially fatal outcomes of xylophagia. Thus,
107 this meta-synthesis was undertaken as an attempt to fill this gap, and provide a better understanding on xylophagia,
108 its treatment, possible causes and postulated relationship with other diseases. A meta-synthesis is a systematic
109 reviewing process that combines and analyses qualitative evidence in a coherent manner, with the primary purpose
110 of developing a deeper understanding of a particular phenomenon (Erwin et al., 2011, Walsh and Downe, 2005,
111 Campbell et al., 2003, Jensen and Allen, 1996). This meta-synthesis was of descriptive type as it analyzed the
112 phenomenon of xylophagia comprehensively.

113 **Methods**

114 A systematic exploration of the qualitative empirical literature on xylophagia was carried out. Our focus was
115 on gaining a better understanding of xylophagia, its prevention, treatment and intervention options, possible
116 etiological causes and postulated relationship with other diseases. Therefore, meta-synthesis approach was used to
117 assimilate and synthesize research studies related to xylophagia.

147 determining how the studies are related; translating the studies into one another; synthesizing translation; and
148 expressing the translation (Hines, 2013, Noblit and Hare, 1988). We began with reviewing every research study
149 various times in order to recognize and record main themes along with details pertaining to the context of the study.
150 We carried out a first order construct which involved making a data table for identifying main ideas and concepts
151 throughout the research articles. This helped in finding the ways in which all research articles were related to one
152 another. We then carried out second-order constructs. In this step, the finding of every research article was
153 compared and contrasted systematically to identify points of convergence and divergence. This also helped us in
154 matching themes across studies and formulating categories of themes. Additionally, we ensured that all the themes
155 paralleled the overall line of argument relating to xylophagia that arose from our review.

156 **Results**

157 A total of 18 articles were included in this meta-synthesis. The characteristics of the 18 cases studies are
158 shown in Table 1. This meta-synthesis comprises 14 case reports, 3 letters to editors and 1 experimental study.
159 Seven studies reported cases of xylophagia from United States of America (US); two studies were carried out in
160 India, one in Iran, Ireland, Germany, United Kingdom, Canada, South Africa, Greece, Australia, and Turkey
161 respectively. There were 16 females and 9 males in included studies. The mean age and standard deviation of males
162 and females were 29.25(12.17) years and 32.81(11.92), respectively. The mean duration and standard deviation of
163 paper pica were 4.80(4.27) years.

164 **Summary of synthesis.** Five primary themes emerged from our meta-synthesis: 1) Precipitation/onset of
165 Xylophagia, 2) Co-morbid psychiatric or medical illnesses, 3) Assessment and diagnostic methods, 4) Outcomes of
166 xylophagia, and 5) Treatment options. The distribution of themes across the 18 papers is detailed in Figure 2. Table
167 2 shows selected data excerpts from included articles.

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169 See Figure 2
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172 See Table 2
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174 **1) Precipitation/Onset of Xylophagia**

175 **Causes and triggers of xylophagia:** Studies included in this synthesis identified various causes and triggers
176 of xylophagia, and in particular modelling of this behavior through vicarious learning (Bharti et al., 2015) including
177 the symptom of celiac disease (Fotoulaki et al., 2007).

178 **Family history:** Some of the family members of xylophagia patient were reported as having a paper-eating
179 habit before their onset (Bharti et al., 2015).

180 **Duration:** Some patients started eating paper or cardboard in order to escape from painful experiences and
181 avoid reality. Duration of consumption was varied, with some reporting more than a year (Chisholm Jr and Martín,
182 1981), two to three years (Bharti et al., 2015, Gowda et al., 2014, Yalug et al., 2007), five years (Callinan and
183 O'Hare, 1988), and 12 years (Olynyk and Sharpe, 1982). There were also adults who started from adolescence
184 (Olynyk and Sharpe, 1982), while others started late in life (Dumaguing et al., 2003).

185 **Maintaining factors:** Various factors were found playing a role in the continuation and maintenance of this
186 behavior. For instance, one study revealed that paper eating alone, learned through instrumental learning, was
187 reinforced and maintained through escape learning. It progressed due to a lack of attention from family members
188 (Bharti et al., 2015). Another study showed that paper eating became a pattern as the patient used to eat paper alone
189 without anyone noticing the odd behavior, thus leading to persistent xylophagia (Gowda et al., 2014). Similarly,
190 distorted beliefs about body image, traumatic family history, the pressure of family responsibility, and lack of
191 amiable relations with a parent played a vital role in onset and continuation of paper eating (Yalug et al., 2007). The
192 habit of eating cardboard also progressed in order to lose weight, control food intake, and attain satiety; as well as to
193 reduce tension and achieve gratification (Callinan and O'Hare, 1988, Stein et al., 1996). Looking as a whole, these
194 findings show an intricate interplay of psychological factors behind increasing vulnerability, acting as precipitating
195 factors as well as maintaining factors of xylophagia.

196 **2) Co-morbidity**

197 **Psychological disorders:** These include inhalant use disorder (Gowda et al., 2014), alcohol dependence
198 (Moore and Sears, 1994), schizophrenia (Klein et al., 2014), obsessive-compulsive disorder (Bharti et al., 2015),
199 intellectual disabilities (Keeling et al., 1987); anorexia nervosa, major depressive disorder, panic disorder with

200 agoraphobia, social phobia, pica, and hair-pulling (Bakhshaeekia et al., 2009, Spaniolas et al., 2010, Stein et al.,
201 1996, Yalug et al., 2007).

202 **Medical diseases:** These include: Mercury poisoning (Olynyk and Sharpe, 1982); anemia (Callinan and
203 O'Hare, 1988); coeliac disease (Fotoulaki et al., 2007); gastritis and duodenitis (Moore and Sears, 1994). These
204 findings reveal that understanding xylophagia is problematic because its symptomatology is complicated by the
205 presence of various co-morbid psychological and medical disorders.

206 3) **Assessment and diagnostic methods**

207 **Medical assessment/laboratory examinations/dietary history:** Individuals with xylophagia present
208 complicated symptomatology which has to be ruled out by a variety of screening and diagnostic procedures. This
209 meta-synthesis reveals that medical specialists employed a complex array of testing procedures, for instance,
210 abdominal x-ray, exploratory laparotomy (Bakhshaeekia et al., 2009); emogram, serum electrolytes, and serum iron
211 tests, stool tests, magnetic resonance imaging, electroencephalography (Bharti et al., 2015); faecal occult blood tests,
212 full blood count, blood film, white blood cell count, serum iron level, liver function test results, levels of serum
213 protein, albumin, calcium, phosphate, urea, and electrolytes, blood glucose levels, and thyroid function
214 measurements (Callinan and O'Hare, 1988). Similarly, diagnosticians have also employed dietary history, hematocrit
215 level test, hemoglobin level test, corpuscular hemoglobin test, corpuscular hemoglobin count test, and serum zinc
216 test (Chisholm Jr and Martín, 1981); Cognitive functions, physical examination, complete blood picture, urine
217 routine and X-ray abdomen, liver function, peripheral blood smear examination (Gowda et al., 2014, Moore and
218 Sears, 1994); and many more (Graham, 1976, Keeling et al., 1987, Klein et al., 2014, Olynyk and Sharpe, 1982,
219 Schmocker et al., 2015, Spaniolas et al., 2010, Stein et al., 1996, Yalug et al., 2007).

220 4) **Outcomes of xylophagia**

221 **Symptoms/psychological problems/health impact:** A number of medical complications were reported, such
222 as bowel perforation, cachectic and malnourishment, disturbed smell and loss of taste senses, abdominal pain,
223 headaches, dyspnea on exertion, chronic fatigue and listlessness, colonic ischemia, peritonitis and intestinal
224 perforation, retinitis pigmentosa, recurrent hypochromic, microcytic anemia, intestinal obstruction, and death
225 (Bakhshaeekia et al., 2009, Bharti et al., 2015, Callinan and O'Hare, 1988, Chisholm Jr and Martín, 1981, Graham,

226 1976, Keeling et al., 1987, Klein et al., 2014, Moore and Sears, 1994, Olynyk and Sharpe, 1982, Schmocker et al.,
227 2015, Fisher, 2014, Spaniolas et al., 2010, Uretsky, 1974). Various psychological problems reported include
228 anhedonia, weeping spells, thoughts about committing suicide, restlessness, anxiety, palpitations, insomnia, loss of
229 appetite, reduction in attention and concentration, anger outbursts, stubbornness, and self-mutilating behaviors
230 (Bharti et al., 2015, Gowda et al., 2014). Quality of life was also impacted, with reports of decreased interest in
231 personal and social life, reduced interaction with family members and friends, reduction of interest in pleasurable
232 activities, poor academic performance, and slacking in household chores (Bharti et al., 2015, Gowda et al., 2014).

233 **Pleasurable effects:** One patient reported that eating paper resulted in feelings of satiety resulting in a
234 reduction in food intake and weight, which then pleased the patient (Callinan and O'Hare, 1988). Similarly, eating
235 notebook paper and newspapers was shown to be the source of immense pleasure (Gowda et al., 2014, Stein et al.,
236 1996). However, this is not the case as one study reported a lack of pleasure from this habit (Bharti et al., 2015).

237 5) Treatment options

238 **Medical care:** Medical treatments recommended by the included studies comprised: prescribing drugs,
239 surgeries or supplements, such as fluoxetine (Bharti et al., 2015); iron dextran intravenous infusion, folic acid and
240 vitamin B₁₂ injections (Callinan and O'Hare, 1988); elemental zinc and ferrous sulphate (Chisholm Jr and Martín,
241 1981); paroxetine, clonazepam, baclofen, supplemental multi-vitamin injectable preparations (Gowda et al., 2014);
242 upper gastrointestinal series, upper endoscopy, iron supplementation (Schmocker et al., 2015); clomipramine,
243 sertraline, citalopram (Stein et al., 1996); olanzapine, fluoxetine (Yalug et al., 2007); and oral ferrous sulphate
244 therapy (Moore and Sears, 1994, Olynyk and Sharpe, 1982).

245 **Psychological care/counselling:** Psychotherapy was employed in four studies, which included
246 psychoeducation for patients and family members, cognitive behavior therapy (CBT) to reduce stress, enhance
247 coping skills, restructure disturbing cognitions, and avoiding damaging behaviors (Bharti et al., 2015, Gowda et al.,
248 2014), overcorrection (Foxy and Martin, 1975), and counselling regarding the disease and appropriate eating
249 behaviors (Schmocker et al., 2015).

250 **Recovery rates and duration:** The findings revealed improvements in symptoms and in some cases patients
251 were reported to be asymptomatic. Improvements in symptoms, no recurrence and aversion towards paper/cardboard

252 eating was seen from a minimum of one week of treatment and ranged to variable time durations of follow-ups
253 (Callinan and O'Hare, 1988, Chisholm Jr and Martín, 1981, Gowda et al., 2014, Moore and Sears, 1994, Olynyk and
254 Sharpe, 1982, Schmocker et al., 2015, Stein et al., 1996). However, one study revealed minimal improvements in
255 symptoms, non-compliance, and aggravation of symptoms after 2 months of hospitalization (Yalug et al., 2007).

256 **Recommendations:** Despite the abnormal nature of this problem, only six research studies suggested
257 recommendations to improve treatment or assessment of xylophagia. Studies have highlighted the absence of
258 standardized medication regimens or psychotherapeutic interventions for treating xylophagia. According to Gowda
259 (2014), support from family, provision of residential care, as well as fostering personal, social and environmental
260 strengths may be beneficial for such individuals. Likewise, another research study highlighted the need for taking a
261 detailed history of individuals presenting with such symptomatology (Spaniolas et al., 2010). Authors also suggested
262 the inclusion of foreign body ingestion for differential diagnosis of abdominal pain (Bakhshaeekia et al., 2009). The
263 need to address nutritional deficiencies for preventing further complications and more research studies to address
264 management options was recommended by three included studies (Schmocker et al., 2015, Stein et al., 1996, Yalug
265 et al., 2007).

266 **Discussion**

267 Our meta-synthesis found that precipitation of xylophagia; co-morbid psychiatric disorder; outcomes of
268 xylophagia; treatment options comprising medical care, psychological care, counselling, and duration of recovery
269 are the most critical points have been discussed. The serious complications of xylophagia include bowel perforation,
270 mercury poisoning, and in one case, death (Bakhshaeekia et al., 2009, Blinder and Salama, 2008, Olynyk and
271 Sharpe, 1982, Klein et al., 2014). Included studies also show a complex interplay of factors that might be involved
272 in xylophagia such as, modelling of this behavior, traumatic experiences during childhood and adolescence,
273 reinforcement and maintenance of this behavior through escape learning, insufficient attention from family
274 members, problematic relations with members of family, and irrational beliefs about body image (Bharti et al., 2015,
275 Callinan and O'Hare, 1988, Fisher, 2014, Gowda et al., 2014, Stein et al., 1996, Yalug et al., 2007). The findings
276 support socio-cultural as well as biopsychiatric factors in etiology, treatment, and prevention of xylophagia
277 specifically, and eating disorders generally.

278 Pica is often reported as a disorder of childhood (Blinder and Salama, 2008, Dumaguing et al., 2003, Klein et
279 al., 2014), making up 25-33% of the cases reported worldwide (Gowda et al., 2014); with incidents rates reportedly
280 decreasing with age. However, our findings revealed that the majority of cases (68%) are adults, with one geriatric
281 patient (Dumaguing et al., 2003). This again points to the underlying cause of pica seen in most of these cases,
282 which is iron-deficiency. There is also the fact that many patients feel ashamed to admit to such behavior (Spaniolas
283 et al., 2010, Bakhshaeekia et al., 2009), a factor also observed in patients in our studies, thus delaying diagnosis until
284 adulthood when severe complications occur. Additionally, children are rarely referred for psychiatric consultations,
285 further delaying the appropriate diagnosis. Thus, Hergüner et al suggested that children should be sent for
286 psychiatric assessments in the absence of underlying medical conditions, OCD, or a family history of OCD; and
287 when iron-deficiency anemia and pica are resistant to iron therapy (Hergüner et al., 2008).

288 **Potential impact of culture and geography**

289 There were a total of 11 countries involved, with the majority of cases reported in developed countries (i.e.,
290 12 respondents) (Callinan and O'Hare, 1988, Fisher, 2014, Graham, 1976, Keeling et al., 1987, Klein et al., 2014,
291 Moore and Sears, 1994, Olynyk and Sharpe, 1982, Schmocker et al., 2015, Spaniolas et al., 2010, Uretsky, 1974)
292 compared to developing countries (i.e., 7 respondents) (Bakhshaeekia et al., 2009, Bharti et al., 2015, Gowda et al.,
293 2014, Stein et al., 1996, Yalug et al., 2007, Fotoulaki et al., 2007). This discrepancy might be a result of more
294 sufficient medical access and knowledge regarding pica as a disorder in developed countries. In a study determining
295 the prevalence rates of psychiatric morbidity in children and adolescents in India where pica is one of the diagnoses,
296 those in urban slum areas had the lowest prevalence rates compared to those in urban areas. This phenomenon might
297 be due to the absence of awareness pertaining to significant psychiatric issues, greater acceptance for deviance,
298 worsened living conditions and the occurrence of various stressors (Srinath et al., 2005). A study by Kettaneh et al
299 also found that being non-European was an independent risk factor for pica, with the authors suggesting that the low
300 incidence in Europeans was either because pica is rare in these patients, or due to the unwillingness to admit to pica
301 because of cultural traits (Kettaneh et al., 2005). As previously mentioned, in some ethnic groups, pica is culturally
302 acceptable, such as in Turkey and Africa where women were encouraged to eat clay to increase their fertility (al-
303 Kanhal and Bani, 1995, Derman et al., 2005).

304 **Underlying etiologies**

305 The majority of patients in the included studies had either iron deficiency or iron deficiency anemia (IDA).
306 This parallels clinical reports as well as findings in the literature that have pointed to the role of iron deficiency, and
307 not necessarily IDA, in the development of pica (Olynyk and Sharpe, 1982, Lumish et al., 2014, Hergüner et al.,
308 2008). Indeed in the study by Foutoulaki et al, the pica was due to IDA secondary to celiac disease (Fotoulaki et al.,
309 2007), which damages the small intestine where iron is absorbed; and is nonresponsive to iron therapy (Freeman,
310 2015). One theory postulated *restoring*, which describes pica as the body's reaction to deficiency (Klein et al.,
311 2014). This then underlines the importance of first excluding and treating underlying and organic causes of pica
312 before initiating psychiatric therapy (Fotoulaki et al., 2007, Dumaguing et al., 2003). There is however debate as to
313 which came first, with some authors arguing that the consumption of non-nutritive substances in itself led to a loss
314 in appetite which then results in iron deficiency. This was seen in the Callinan paper where cardboard chewing was
315 believed to render fullness which in turn resulted in reduced food intake, consequently impairing iron absorption
316 (Callinan and O'Hare, 1988). It is also postulated that the consumption of these materials may have resulted in an
317 inhibition of iron absorption, thus leading to iron deficiency (Moore and Sears, 1994). Nevertheless, in most of the
318 cases studied in the present meta-synthesis, individuals with iron-deficiency were cured or at least showed
319 improvement in xylophagia after treatment with iron supplementation. However, although a possible relationship
320 between pica and iron-deficiency has been shown, for now, the mechanism is still unclear (Callinan and O'Hare,
321 1988, Chisholm Jr and Martín, 1981, Moore and Sears, 1994, Olynyk and Sharpe, 1982, Schmocker et al., 2015).

322 A few patients had intellectual disabilities, which supports previous reports that pica is more prevalent in
323 patients with mental disabilities, including having a higher mortality rate (Dumaguing et al., 2003, Klein et al., 2014,
324 Hergüner et al., 2008). Indeed it was noted that worldwide 10-15% of those with pica have learning disabilities
325 (Gowda et al., 2014). It has been postulated that this is due to diminished impulse regulation, which is a critical
326 clinical characteristic in pica (Gundogar et al., 2003, Stein et al., 1996). The Stein patient had a comorbidity of OCD
327 (Stein et al., 1996), while other patients reported symptoms of compulsions (Schmocker et al., 2015, Yalug et al.,
328 2007, Moore and Sears, 1994); which again parallels previous findings which have attributed pica to OCD or
329 obsessive-compulsive traits (Gundogar et al., 2003, Dumaguing et al., 2003, Bharti et al., 2015). Indeed in instances
330 where iron therapy failed, the use of a selective serotonin reuptake inhibitor was shown to be effective (Bharti et al.,
331 2015, Gowda et al., 2014, Yalug et al., 2007). Other reported predisposing factors include schizophrenia (Gowda et

332 al., 2014, Hergüner et al., 2008), seen in two cases (Dumaguing et al., 2003, Klein et al., 2014); as well as those with
333 a family history of pica, also seen in two cases (Schmocker et al., 2015, Bharti et al., 2015).

334 **Recommendation for early prevention**

335 Early detection of xylophagia can help in preventing complications (Singh et al., 2015). As primary
336 prevention, pica should be screened by conducting prevalence surveys, direct observation, stool checks, medical
337 history review, interviewing caregivers, and using screening scales such as the Behavior Problem Inventory in
338 clinical programs and residential facilities (Williams and McAdam, 2012). Those who work where they will interact
339 with people with pica should also be taught about what pica is, the dangers of ingestion of such non-food substances
340 and the prevention strategies (Williams et al., 2009). Family members can also play a role by paying closer attention
341 to patient's behavior, and seeking medical advice immediately if any unusual habits are observed. Parents should
342 also periodically check for any tooth deterioration that might be a result of pica behavior (Singh et al., 2015).

343 **Implications for future research, policy and practice**

344 This meta-synthesis shows that there are many challenges associated with diagnosing pica because of limited
345 data on its characteristics, medical and psychological assessments, and management. Patients also often present with
346 nonspecific signs and symptoms which may make it hard to detect as well as lead to misdiagnosis and unnecessary
347 treatment (Bharti et al., 2015). This is aggravated by the fact that most patients conceal this behavior and seek
348 medical assistance only after complications occur (Spaniolas et al., 2010), as seen in three cases (Bharti et al., 2015,
349 Schmocker et al., 2015, Graham, 1976); or a proper history could not be obtained, especially in cases of intellectual
350 disability or children (Bakshaeekia et al., 2009). Various biopsychiatric and sociocultural components have been
351 shown in this review (i.e., familial factors, maintaining factors, co-morbid conditions, psychological problems as
352 outcomes and treatment options) which support the notion that these aspects operate simultaneously in additive,
353 multiplicative, and/or transactional ways. Thus by being cognizant of socioeconomic factors, as well as detecting
354 xylophagia in the early stages, prevention can be achieved and complications can be avoided. More clinical
355 examples are also required to analyze whether pica is more prone to happen in developing or developed countries, as
356 there is a possibility that the cases in developing countries are not reported because of poor access to medical
357 resources and/or naivety towards the characteristic of pica.

358 Currently, the treatment for xylophagia is limited, and in most cases, patients are treated with oral iron
359 therapy, antidepressants, or by surgically removing the intestinal bezoars. From a medico-legal perspective, the
360 focus of treatment should be on the main disorder and correcting the causal deficiencies or emphasizes the
361 complication (Moore and Sears, 1994). In some cases, a combination of antipsychotic medication and cognitive
362 behavior therapy is opted (Knecht, 2000).

363 Overcorrection procedures have also been used, but only with the institutionalized patients. The fact that pica
364 is associated with obsessions and compulsions also suggest the need for behavioral treatment (Dumaguing et al.,
365 2003). Further studies, however, are still warranted to investigate the effectiveness of other possible treatments. As it
366 pertains to the substance consumed, it is important to gather information on the specific type that was consumed in
367 terms of brand names, as different brands of the same product may contain different materials, which could lead to
368 different outcomes. In the same vein, while it may all come under the umbrella term of xylophagia, consuming
369 pages of a book would have different consequences from consuming toilet paper. Thus explored and collected
370 information collected should be as specific and detailed as possible (Young et al., 2008).

371 The unusual intake of foreign inedible substances by medical imaging and post-mortem examination with
372 concurrent non-existence of role of external party or influence of medication, illegal drugs, or intoxicating
373 substances, allows an effective explanation of the findings in the background of a complicated biopsychosocial
374 clinical picture. As pica is not commonly known and often asymptomatic, patients might continue practicing the
375 habit until complications occur. As such, there should also be a concerted effort to promote the awareness of pica
376 and its possible consequences to both the public and health professionals. More well-designed trials should also be
377 carried out to understand the characteristics and risk factors of xylophagia in order to facilitate early detection and
378 prevention, as well as diagnosis.

379 **Limitation**

380 There are some limitations to this meta-synthesis. One limitation was the lack of in-depth exploration of the
381 phenomenon in the included studies. Studies did not explain the results of psychological assessment in details, and
382 there was an absence of follow-up monitoring to confirm the efficacy of treatments offered. Furthermore, the term
383 “xylophagia” was rarely used in the articles; instead, the specific substance consumed was mentioned, possibly
384 increasing the difficulty in retrieving related studies as xylophagia encompasses a wide range of substance that can

385 be consumed. Out of 18 studies included in this meta-synthesis, 15 studies were case reports, showing a dearth of
386 empirical studies with detailed qualitative or mixed-methodology to highlight the underpinnings of xylophagia.

387 **Conclusions**

388 The commonly reported causes and triggers of xylophagia are behavior modelling through vicarious learning
389 and post-traumatic disorder suffered during childhood and adolescence. It has been found that xylophagia is often
390 associated with other diseases such as iron-deficiency and various psychiatric disorders. A positive effect of patient
391 and family members undergoing psychology counselling or cognitive behavior therapy in reducing stress and
392 enhancing coping skills thus avoiding self-damaging behaviors has been established. However, out of the 18
393 included studies, only four employed psychotherapy. Similarly, standardized medication regimens for treating
394 xylophagia are still not available and further research is still required to better understand the disease. Furthermore,
395 there must be a thorough assessment of medical, conceptual and explorational histories for any indication of pica
396 that may assist in post-mortem examination and may also give relevant details of linked medical and psychiatric
397 problems. This thorough assessment can help to overcome the chances of overlooking or misdiagnosis due to the
398 presence of transient or mild symptoms or the occurrence of overlapping symptoms.

399 **Compliance with Ethical Standards**

400 **Disclosure of potential conflicts of interest**

401 All authors have no conflict of interest.

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